

## Case Report

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# Brain metastasis of thyroid adenoid cyst carcinoma: Unusual case report

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## Abstract

**Background:** Cerebral metastasis from Adenoid cyst Carcinoma (ACC) remains extremely rare.

**Case description:** 62-year-old female, with a history of thyroidectomy for goiter surgery associated with left cervical lymphadenopathy five years ago. She presented a decrease in visual acuity at 6/10 in the right eye. Brain MRI revealed an intra-conical right orbital lesion process associated with a right frontal cerebral lesion process, three other osteolytic lesions of the cranial vault, lung metastases, and hepatic metastasis. Thyroid ACC with Antibody Anti CD117+ after stereotaxic biopsy of brain lesion was retained by the pathologist. She underwent radiotherapy and chemotherapy management. At 6 months follow-up, the cerebral and liver localisations didn't progress, unfortunately, the lung mass was progressed from 4 cm to 6 cm.

**Conclusion:** Multiple brain metastasis from ACC of the thyroid is extremely rare. Diagnosis and Management could present some challenges for surgeons and oncologists.

**Keywords:** Adenoid cyst carcinoma; Thyroid; Brain metastasis; CD117; Stereotaxic biopsy.

## Introduction

Cerebral metastasis from Adenoid Cyst Carcinoma (ACC) remains very rare [1]. Authors reported Bartholin's gland, Breast, Lung, parotid adenoid cystic carcinoma (ACC) with metastasis to the brain [2-5]. The multiple metastatisation patterns—nodal are correlated with poor prognosis [2]. However, the diagnosis may encounter some challenges when a primitive lesion was not known.

We present a rare case of multifocal cerebral metastasis from thyroid ACC with other secondary localization such as liver and lung involvement.

## Case description

### History and clinical presentation

62-year-old female, with a history of thyroidectomy for goiter surgery associated with left cervical lymphadenopathy five years ago, without any other medical diseases. We do not find any pathology reports before because the patient was not managed in our department.

The history of the disease is marked 6 months before its admission by the progressive installation of a decrease in visual acuity at the right eye associated with headaches, and vomit-

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ing motivating a consultation with an ophthalmologist and a neurosurgeon. The clinical examination at admission shows a conscious patient GCS at 15, without cognitive disorders, with a decrease in visual acuity at 6/10 at the right eye without sensory-motor deficit. ophthalmological examination shows a right central scotoma, without eye proptosis. Local skin examination was normal.

### Diagnosis assessment

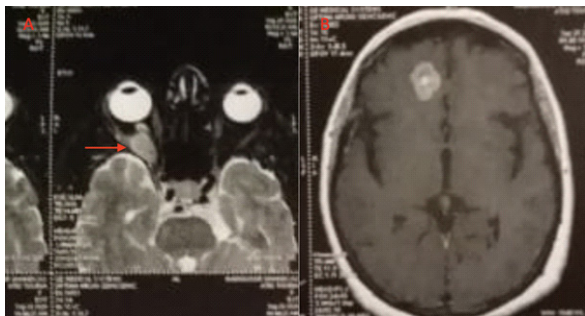
The sample's blood exam for the hormonal function was normal, under Levothyrox treatment.

Brain Magnetic Resonance Imaging (MRI) was performed and revealed an intra-conical right orbital lesion process filling the orbital apex and laminating the optic nerve, associated with a right frontal cerebral lesion process (Figure 1) and three other osteolytic lesions of the cranial vault evoking a priori secondary locations (Figure 2). In addition, Thoraco-abdomino-pelvic Scan reveals multiple lung metastases and hepatic metastasis.

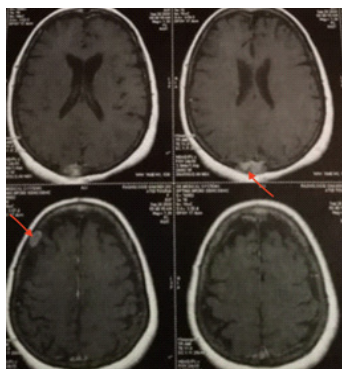
The morphological and immunohistochemical confirmed a cerebral metastasis from thyroid adenoid cystic carcinoma with Antibody Anti CD117+ (Figure 3).

### Medical management

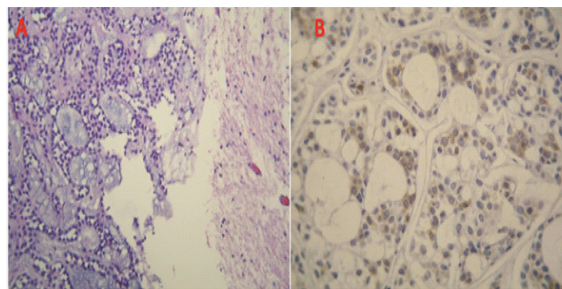
Corticosteroids such as intravenous methylprednisolone 120 mg/day for one week then prednisolone 20 mg tablet for 3 weeks (60 mg per day for one week, 40 mg per day for another one week, then 20 mg per day for another last week). Anti-epileptic such as valproate of Sodium 1500 mg per day were started for 6 months initially. Antalgic such as paracetamol 1000 mg every 6 h in a case of headaches.



**Figure 1:** A: Intra-conical right orbital lesion process filling the orbital apex and laminating the optic nerve, associated with a right frontal cerebral lesion process.



**Figure 2:** A: Osteolytic lesions of the cranial vault evoking a priori secondary locations (Red Arrow).



**Figure 3:** A: HEX100, shows Infiltration of the cerebral parenchyma by a round cell tumor proliferation with a cylindrical appearance), B: Immunohistochemistryx200: tumor cells express Anti CD117 Antibody.

The patient received adjuvant cerebral X-Ray therapy with chemotherapy for the lung secondary mass progression.

### Surgical management

Stereotaxic biopsy under sedation and local anesthesia was performed for the frontal intracerebral lesion for histology and Immuno-histochemical.

### Follow-up

The outcome at 6 months was good with the improvement of visual acuity at 8/10, no neurological deficit, and stability of cerebral and liver localisations without progression, unfortunately, the lung mass was progressed from 4 cm to 6 cm.

### Discussion

We report an unusual case of multifocal cerebral metastasis from Thyroid Adenoid Cyst Carcinoma with other secondary localization such as liver and lung involvement.

Adenoid cyst carcinoma was reported for the parotid gland, Bartholin's gland, and Breast, the metastatisation are been reported for the Brain, the kidney, lung, and pituitary [1-7]. The most common secondary location of thyroid ACC is the lung (20%) followed by the bones (4%) and the liver (3%), the brain location is scantily reported [8,9].

ACC thyroid may well mimic goiter or upper airway tract tumor invading the thyroid gland [10,11].

In our case, it was thyroid ACC associated with lymphadenopathy treated first by Thyroidectomy which progressed five years later with multifocal cerebral metastasis with liver and lung metastasis. 10 cases with female (60%) predominance and an average range of 49.8 years were reported before in the literature and none of the cases presented a brain metastasis [12].

The primary treatment for metastatic ACC is surgical resection followed by adjuvant radiotherapy [1], however, for multiple brain metastasis as in our current case, we preferred to make a stereotaxic biopsy of accessible brain lesion to confirm the diagnosis and send the patient for radiotherapy and chemotherapy for the lung progression mass.

Thus we think that in our current case the diagnosis must be done 5 years ago and the diagnosis of a simple goiter of thyroid might be wrong. The right diagnosis is based on the immunohistochemical staining profile as follows: Alcian blue (AB) (+), cyto-keratin (CK) 5/6 (+), CK8/18 (+), CD117 (+), thyroid transcription

factor-1 (TTF-1) (–), thyroglobulin (Tg) (–), chromogranin A (CgA) (–) [8]. In our case, the immunohistochemical revealed expression of the Anti CD117 Antibody by the tumor cells.

Authors reported that ACC has an excellent prognosis, contrary to the extra-Breast form (5-year survival is almost 90% for the former and only 30%–50% for the latter) [13], this might explain that five years later our patients progress with multifocal metastatisation such as brain, liver, and lung. Unfortunately in our case, the lung metastasis progressed, however, the brain metastasis was stable with no new brain lesion after radiotherapy.

### Conclusion

Brain metastasis from Thyroid ACC is extremely rare. Diagnosis and Management could present some challenges for surgeons and oncologists. Besides Management of a metastatic situation presents more challenges. However, a discussion case by case should be mandatory between surgeons and oncologists. In the case of multiple brain metastasis, a biopsy for diagnosis should be done before oncology treatment, and the other metastasis extra cerebral should be taken into account in the management by the surgeons and the medical oncologist.

### Declarations

**Consent for publication:** NA

**Competing of interest:** None.

**Acknowledgment:** none.

**Authors Contribution:** MH: case management, supervision, reviewing, and editing; NDAB: conceptualization, writing original draft, writing, reviewing, editing; OB: reviewing and Editing; SS: pathology analysis, reviewing and editing, NEF: Validation, Editing; MREM: Validation, supervision.

**Financial disclosure statements:** None.

**Conflict of interest:** The authors do have not any conflicts of interest in this case report and any financial resources.

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